Letters to the Editor

Idiopathic focal myositis in pregnancy

Sir, Focal myositis is a rare, benign inflammatory disorder of skeletal muscle, the cause of which is unknown [1]. It usually affects limb musculature, although rarely muscles of the head and neck [2] and spine [3] can be involved. The differential diagnosis is wide and includes more sinister lesions such as pyomyositis and soft tissue tumours. Early histological diagnosis is therefore essential. The development of the more common generalized inflammatory myopathies, polymyositis and dermatomyositis, has been reported in pregnancy [4]. The subsequent clinical course in these disorders is variable. We report a case of idiopathic focal myositis affecting the abductors of the thigh developing acutely in a young woman during pregnancy, with subsequent spontaneous remission.

A 32-yr-old woman presented with a 3-week history of pain in her right groin. She was 11 weeks pregnant. This was her first pregnancy which was otherwise uncomplicated. There was no other significant past medical history. The pain was progressively worsening such that she could no longer walk unaided. There was no history of injury and she had no other symptoms.

![Fig. 1](A, B) Axial and coronal T1-weighted MRI through the right thigh demonstrates low signal intensity and loss of normal textural pattern of flexural and abductor muscle groups (arrows). (C) Corresponding coronal T2-weighted image shows diffuse increased signal intensity in gluteus minimus, rectus femoris and the insertion of ileopsoas, this representing proliferative myositis (arrowhead).
On admission she was taking paracetamol and folic acid. No other medication had been taken during the pregnancy.

On examination she was apyrexial but distressed. There was no rash or lymphadenopathy. The right hip was held flexed on lying supine. Slight swelling was seen over the right greater trochanter with an area of overlying erythema. The anterolateral aspect of the upper thigh and the right groin were excessively tender. All movements of the right hip were painful, particularly adduction and external rotation. The remainder of the musculoskeletal examination was normal.

The investigation revealed: haemoglobin 11.7 g/dl, white cell count 6.89 × 10⁹/l (normal differential), platelets 257 × 10⁹/l, erythrocyte sedimentation rate (ESR) 59 mm/h and C-reactive protein (CRP) 65 mg/l. The aspartate aminotransferase was 19 U/l (normal 14–36) and creatine kinase (CK) 148 IU/ml (normal 30–106). Alkaline phosphatase, calcium and electrolytes were normal. Plain X-rays of the hips were normal. Magnetic resonance imaging (MRI) of the thighs and hip joints inflammatory lesion of the muscles of the extremities. This showed a diffuse area of increased signal intensity around the right hip involving the surrounding muscles, which appeared oedematous (Fig. 1). There was increased signal in part of the glutaeus minimus, the origin of rectus femoris and the insertion of the ilopsoas muscles. The right hip joint appeared normal; there were no signs of osteonecrosis.

Surgical exploration of the right hip was performed by an anterior iliofemoral approach. Macroscopically the femoral head, acetabulum and joint capsule appeared normal. Synovial fluid microscopy showed scanty polymorphs only. Incisional biopsies of the deep abductors of the hip showed preservation of the fascicular architecture. A focal, predominantly perivascular, chronic inflammatory cell infiltrate was seen, but no specific diagnostic features were identified (Fig. 2). Eosinophils or granulomata were not seen. Biopsies of the superficial muscle group were normal, but those of fat also showed a perivascular chronic inflammatory cell infiltrate, in addition to degenerative changes. Micro-organisms were not seen on microscopy and cultures of tissue specimens were sterile. Blood cultures were also sterile and coxsackie virus group B neutralization tests were negative. She was antinuclear antibody (ANA)-negative and immunoglobulin levels were normal. Electromyography was not performed.

This patient presented with an acute localized non-suppurative myositis in pregnancy, that spontaneously improved without specific therapy. MRI demonstrated the focal nature of the disease. Pyomyositis and more generalized myopathies were excluded, as was a muscle neoplasm. Focal myositis was first described by Heffner et al. [1], their series of cases beginning as a benign inflammatory lesion of the muscles of the extremities. Focal myositis affecting the tongue [5] and sternoclavido-mastoid muscles [2, 6] has also been reported, the macroscopic appearance of which coined the term ‘pseudotumour’. The histology in the first series of cases was one of a lymphocytic infiltrate, interstitial fibrosis and occasional muscle fibre necrosis. No gender difference in incidence was reported and the median age at diagnosis was 39 yr. A viral aetiology has been suggested [7]. The course of the disease is usually benign, often with spontaneous resolution [2]. Corticosteroids have been required in some cases, however, usually with good results [3]. Obtaining a histological diagnosis is essential to avoid unnecessary surgical treatment.

Polymyositis may begin as a localized lesion [8, 9]. In this case there has been no clinical evidence of progression to a generalized myopathy. Polymyositis and dermatomyositis developing during pregnancy are associated with a variable outcome [4]. Evidence suggests an adverse outcome in terms of prematurity, abortion and perinatal death. To our knowledge, focal myositis has not been reported in pregnancy and fortunately in this case it was not associated with an adverse effect on the fetus. If this disorder represents a self-limiting subtype of polymyositis then its appearance in pregnancy might not be simple coincidence. However, this appears to be a separate disorder with a more favourable prognosis. Its appearance in pregnancy might suggest a hormonal influence on an unidentified aetiological agent, although the rarity of the condition makes further evaluation of this extremely difficult.

The treatment of inflammatory myopathies in pregnancy is complicated by the risks associated with the use of corticosteroids and other immunosuppressive drugs. Fortunately such agents were not required in this case. Other uncommon causes of hip pain in pregnancy include transient osteoporosis of the hip and avascular necrosis. Although rare, idiopathic focal myositis should be considered in the differential diagnosis of hip pain in pregnancy. This case also emphasizes the need to exclude

Fig. 2. Muscle biopsy. Focal perivascular chronic inflammatory cell infiltrate.
carefully other lesions that might have serious bearing on mother and fetus.

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